



## Idiopathic granulomatous mastitis: A diagnostic and therapeutic challenge



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### ABSTRACT

**Background:** Idiopathic granulomatous mastitis is a rare benign breast disease of women of reproductive age. It usually presents as a painful mass. Since the etiology is unclear, directed diagnosis and management is lacking.

**Methods:** This is a retrospective chart review of 14 patients, over twelve years (2004–2016), identified through query of pathology findings.

**Results:** Two asymptomatic patients were diagnosed after oncologic breast resection following neo-adjuvant chemotherapy. The remaining twelve patients were young (31.7 years, range 23–43 years), predominantly non-white (50% African/African-American, 36% Hispanic, 7% Asian), pregnant within the last five years (86%), with no prior granulomatous disease. Evaluation included breast imaging, microbial cultures and staining, and biopsy. Treatment included antibiotics (57%), corticosteroids (21%), methotrexate (7%), and/or surgery (71%). Imaging suggests segmental masses, possibly abscess.

**Conclusion:** Granulomatous mastitis is uncommon, and difficult to diagnose and manage. We review our experience, the literature, and propose an algorithm for diagnosis and management.

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## 1. Introduction

Idiopathic granulomatous mastitis is a rare benign inflammatory breast disease first described in 1972 by Kessler and Wolloch.<sup>1,2</sup> The most common presenting symptom is a unilateral, firm, and discrete breast mass which may be accompanied by overlying skin changes and/or possible lymph node involvement with idiopathic granulomatous mastitis.<sup>3</sup> The lesion may occur in any quadrant of the breast, but often extends radially from the retroareolar region.<sup>4</sup> The disease often presents in women of childbearing age with a recent history of pregnancy or ongoing lactation.<sup>5</sup> The overlap of

presenting symptoms with other disease processes such as malignancy, acute or chronic infections, and chronic inflammatory diseases makes definitive diagnosis difficult.<sup>1,6,7</sup> Because the differential may include malignancy, the patient may experience significant anxiety during the evaluation.<sup>8</sup> In addition, the broad differential and the lack of pathognomonic features make definitive diagnosis difficult, often resting as a diagnosis of exclusion on a clinical basis. Because diagnosis is difficult, patients typically have received prolonged courses of antibiotics, frequent biopsies, or surgical procedures. Once the diagnosis is made, treatment strategies are not clearly delineated but are often supportive.

We report a twelve-year experience with the diagnosis, treatment, and management of idiopathic granulomatous mastitis in an academic medical center. Fourteen patients with clinical follow-up were identified using a query of pathology reports. Given findings at presentation, we propose an algorithm for management, with a discussion of its rationale, including radiologic testing, diagnosis including pathology, and management of idiopathic granulomatous mastitis.

**Abbreviation:** IGM, idiopathic granulomatous mastitis; mg, milligram; NSAIDs, non-steroidal anti-inflammatory drugs.

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## 2. Methods

Prior to initiating the study, we obtained its approval from the Institutional Review Board. A retrospective chart review was performed on patients who had undergone breast biopsy and were found to have the pathologic diagnosis of granulomatous mastitis at our medical center over a 12-year period (2004–2016). Each patient's chart was reviewed for pertinent demographic data including age, ethnicity, recent pregnancy, symptoms and duration, as well as medical comorbidities that may include a history of any of the following: tuberculosis, sarcoidosis, chronic inflammatory or granulomatous diseases.

Results of microbial cultures or stains, imaging (mammography, ultrasound, and/or MRI) and biopsy results were noted. Perioperative records, if present, were reviewed, and details were collected including type of surgery and its extent. The medications used in management were documented including antibiotics, corticosteroids, and methotrexate.

## 3. Results

### 3.1. Demographics and risk factors

Using a query of pathology results, 17 patients were identified with the initial query of pathologic findings of granulomatous disease in breast biopsies. Three patients had not sought clinical management after image-guided biopsies; we had no clinical records regarding further evaluation or treatment, and efforts to contact them failed. Of the fourteen evaluable patients, the mean age was 31.7 years and all within reproductive years (Table 1). The high majority were of Hispanic or African/African-American ancestry. No patient had a history of previous granulomatous disease (tuberculosis, sarcoidosis, autoimmune disease, or granulomatous disease). Twelve of the fourteen patients had been pregnant within the last five years.

### 3.2. Presenting symptoms

Two patients were asymptomatic and were included in the study because of the presence of granulomatous mastitis found incidentally in pathology specimens obtained during oncologic procedures. Of the remaining twelve patients, the average duration of symptoms was 3.9 months with the most common signs and symptoms including discrete mass, tenderness to palpation, erythema, and swelling (Table 1). The pain could be out of proportion to findings, suggestive of a localized ischemic etiology. The pain was the motivating factor in prompting all symptomatic patients to seek consultation.

### 3.3. Evaluation

Imaging included chest roentgenography to better evaluate for possible sarcoid, mammography, and ultrasonography. Figs. 1 and 2 demonstrate some of the more typical mammographic and ultrasonographic findings of granulomatous mastitis: segmental masses with ill-defined margins, with tubular structures extending from the mass, which is heterogeneously hypoechoic. The majority (71%) of patients had both aerobic and anaerobic cultures sent on aspirated fluid, half the patients had mycobacterial cultures sent and/or acid fast staining. Three aerobic cultures were positive; two grew *Corynebacterium* species and the other grew *Propionibacterium acnes*—all skin flora. Core needle biopsies were obtained in 79% of patients for diagnosis; the remainder were by excisional biopsy.

**Table 1**

Summary of 14 patients with histology consistent with idiopathic granulomatous mastitis.

Mean Age in years (range)	31.7 (23–38)
<b>Race</b>	
White	7%
Asian	7%
Hispanic	36%
African American	50%
Duration of Symptoms in months (range)	3.9 (0.5–16)
Prior Diagnosis of Granulomatous Disease	0
Pregnancy within last five years of diagnosis	86%
<b>Presence of Symptoms or Signs</b>	
Mass	100%
Pain or tenderness	79%
Erythema	50%
Swelling	29%
<b>Imaging Evaluation:</b>	
Chest x-ray	57%
Mammogram	71%
Ultrasound	79%
<b>Biopsy</b>	
Core/Incisional	79%
Excisional	21%
<b>Systemic Treatment</b>	
Antibiotics	57%
Steroids	21%
Methotrexate	14%
<b>Surgical Intervention</b>	
Wide local excision/partial mastectomy	64%
Total mastectomy	7%
<b>Follow-up</b>	
Interval followed	3 months
Time from diagnosis to symptom resolution	2–24 months

### 3.4. Treatment

The majority (62.5%) of patients were given a trial of antibiotics, most commonly with a short (2 week) course of beta-lactamase resistant penicillin, assuming the more common mastitis at initial presentation. With subsequent diagnosis and a lack of culture growth, three patients were treated with prolonged course of low dose corticosteroids (Table 2) in an effort to spare them surgical resection, as there was data to support this process and the patients preferred to avoid surgery. Of these patients, two of the three patients failed therapy; one of these two patients developed intolerable Cushingoid side effects by two months, leading to discontinuation of therapy, and one did not have symptom improvement over three months. These two were subsequently switched to methotrexate, but the second patient who had no Cushingoid side effects did not tolerate the side effects of methotrexate. This was a patient who had previously undergone partial mastectomy for diagnosis and who recurred within two months. She elected to undergo total mastectomy 23 months after her first procedure. The third patient initially treated with steroids was successfully treated; the corticosteroid was discontinued because of a second pregnancy. Nine patients (64%) underwent surgical procedure, most often partial mastectomy, either for symptom control or for diagnosis.

## 4. Discussion

Our retrospective clinical study found that patients presenting with symptomatic idiopathic granulomatous mastitis are young, usually non-white women of reproductive age, typically with a recent pregnancy within the last five years. The two women who had not been pregnant within the prior five years before diagnosis

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